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Agenesis of the Lung and Contralateral Diaphragmatic Hernia in a Newborn Infant

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CONGENITAL ABSENCE OF A LUNG is a relatively rare anomaly. It is compatible with normal development and longevity. Congenital defects associated with it frequently cause death in the neonatal period. In the case presented here, congenital right pulmonary agenesis was associated with left Morgagni foramen diaphragmatic hernia in a newborn child. The baby lived after surgical repair of the hernia. No similar case has been described in the literature.

REPORT OF A CASE

The patient, a 6 pound, 3 ounce girl, was born by outlet forceps delivery with episiotomy. It was noted that the feet remained cyanotic during the first day of life. An incubator was used and oxygen was given. The infant was discharged the next day, with the mother, the color being reported as normal.

Two weeks later she was readmitted to the hospital with a history of coryza for a week and increasingly difficult breathing and cyanosis for the preceding few days. Feeding had also been poor.

On examination, slight cyanosis and noisy respiration was noted. There were diminished breath sounds over the right side of the chest. The trachea was midline. There was questionable retraction of the chest on the right side, and the heart sounds were heard on the right.

Postero-anterior and right lateral films of the chest (Figure 1) showed opacity on the right, with the heart retracted to that side. At the left cardiophrenic angle there was a cystic, air-containing area which apparently was bowel above the diaphragm.

With the patient under oxygen-ether anesthesia, bronchoscopic examination was carried out with the 3-16 Jesberg bronchoscope. The main carina was observed to be rotated 30 degrees clockwise. The right main stem bronchus ended in a blind stump about 0.5 cm. in length. After the bronchoscope was removed an endotracheal catheter was inserted and the infant's condition became precarious, oxygenation being carried out by quite forcefully squeezing the anesthesia bag.*

A midline upper abdominal incision was made and a defect two and a half inches long in the anterior part of the left diaphragm, in the usual area of the foramen of Morgagni, was observed. A peritoneal sac was present. The defect was closed with interrupted silk imbricating the sac. The pleural cavity was not opened. As the defect was closed the pronounced paradoxical motion of the diaphragmatic defect was corrected and the infant's condition immediately improved. The abdominal wall was closed in layers with silk.

The postoperative course was uneventful. Oxygen was given for four days, aerosol being used for humidification. An x-ray film of the chest showed no hernia. The patient was discharged four days after the operation. She developed normally but had frequent episodes of respiratory tract infection which necessitated hospitalization five times between the sixth and the eleventh month of life. On each occasion the disease was accompanied by difficult breathing and wheezing, and in spite of administration of antibiotics and inhalation of steam at home, invariably progressed to bronchiolitis with cyanosis. Oxygen-

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*Dr. Paul Pentecost administered the anesthesia.

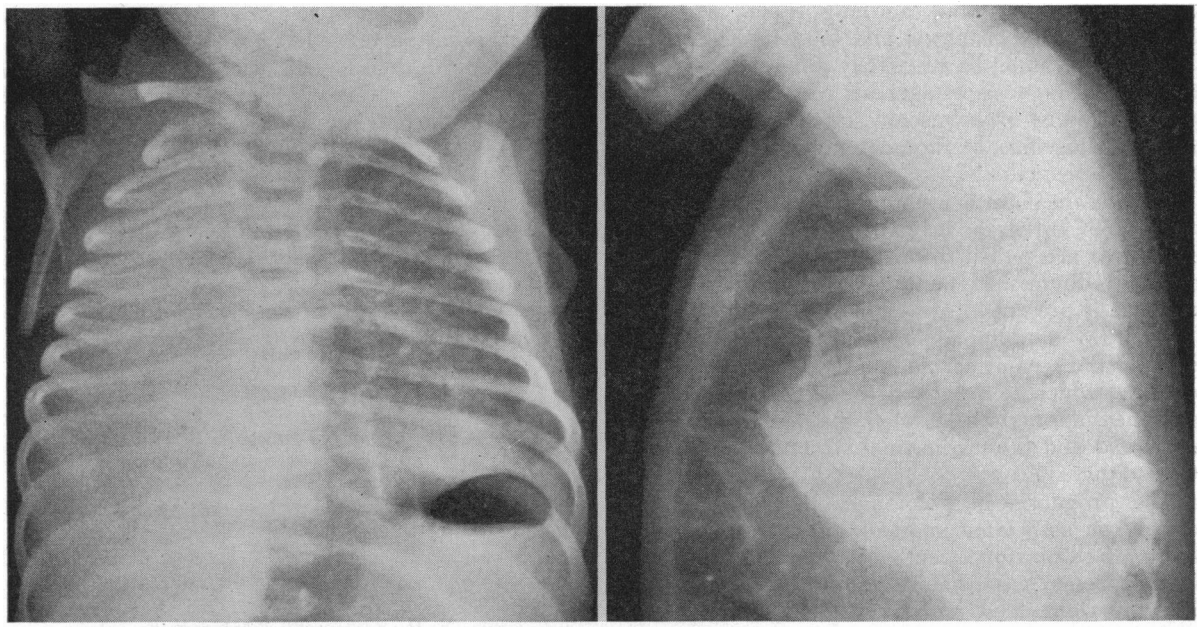

Figure 1.—X-ray films showing herniation of the bowel through the anterior portion of the diaphragm and the absence of the right lung.

aerosol therapy was given each time and the patient recovered in three to four days. As she grew older the attacks of respiratory tract disease became less severe. When free of infection she was alert and active. The growth pattern was within normal limits. Figure 2 is an x-ray film of the chest taken two years after the operation.

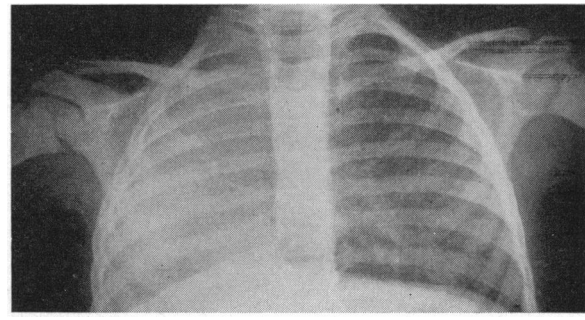

Figure 2.—Diaphragm normal two years after repair of the hernia.

DISCUSSION

Congenital absence of the lung has been reported with increasing frequency in recent years. Valle in January, 1955,[5] reviewed the literature and assembled data on 120 cases, about half of which were reported the 12 previous years. The oldest patient of record died at the age of 72 years. Although the absence of a lung might conceivably decrease a patient's ability to resist respiratory infections, survival in this condition depends mainly on the presence of other congenital anomalies. Despite the frequent association of other anomalies with pulmonary agenesis, there was only one other diaphragmatic anomaly mentioned in the literature. This was in the case, reported by Hanson[2] in 1901, of an infant surviving only about 15 minutes with agenesis of the left lung, which at autopsy "looked like a bunch of millet seed not larger than a buck shot," and a rudimentary left diaphragm consisting of a "small band of muscular fibers about one-eighth inch in width along the border of the ribs." Posteriorly there was no diaphragm at all in this case, the left pleural cavity being filled with small and large bowel. The malrotated cecum and appendix lay at the apex against the clavicle.

Campanole and Rowland[1] reported five cases of hypoplasia of the lung associated with ipsilateral Bochdalek foramen hernia. At autopsy the weight of the lungs was 5.5 grams in two cases, and in the three others 2.5, 5 and 6 grams. In a case of ipsilateral Bochdalek foramen hernia and hypoplasia reported by Tolin,[4] normal aeration of the hypoplastic lung eventually occurred.

These cases apparently represent a different condition than that reported by Hanson, in that the major bronchi were present and the lung tissue was capable of aeration. In Hanson's case, unlike the case herein reported, the hernia was on the same side as the agenesis. Also in the present case the diaphragmatic defect was not as large and a peritoneal sac was present, hence respiratory embarrassment was less severe. However, the paradoxical motion of the herniated abdominal contents through the anterior defect did cause considerable difficulty during anesthesia. Certainly the combination of anomalies in the present case would not have been compatible with prolonged life had not correction of the hernia been carried out.

Pathologically, Schneider[3] has classified agenesis into: (1) True aplasia, (2) hypoplasia with rudi-

mentary bronchus and no lung tissue, and (3) hypoplasia with bronchus and lung tissue present. These types cannot be accurately differentiated clinically. The diagnosis of agenesis rests during life on examination by bronchoscopy, by bronchograms, by angiocardiograms, by thoracotomy or by a combination of these methods.

Usually the chest is asymmetrically flattened, with respiratory lag on the involved side. The mediastinal structures are pulled toward the involved side and signs of diminished aeration on the affected side are usually present.

By x-ray a radiopaque hemithorax is noted, except for the area of herniation of the lung across the midline, which is usually present. On the involved side the diaphragm is elevated, the intercostal spaces narrowed and the mediastinal structures are pulled toward that side.

The prognosis in pulmonary agenesis depends mainly on associated anomalies. The repeated episodes of respiratory tract disease in the early life in the present case, before growth of the left lung into the right chest had taken place, illustrate the added danger of infections in such patients, owing to reduced ventilatory reserve. Since usually in cases of diaphragmatic hernias of the foramen of Morgagni there is a peritoneal sac, the transperitoneal approach is far superior to the thoracic approach for repair of the defect. This was particularly true in the present case, for it obviated pneumothorax on the side of the only lung.

SUMMARY

A patient with congenital absence of the lung and contralateral diaphragmatic hernia—a combination of anomalies not previously reported—was treated successfully by repair of the hernia.

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